



QUALITY OF LIFE IN DYSPHONIC CHILDREN MEASURED ON PEDIATRIC VOICE-RELATED QUALITY OF LIFE (PVRQOL) SCALE IN SERBIA

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SUMMARY – Hoarseness occurs in children of both genders, from the earliest age and beyond, and is caused by improper use or overuse of vocal apparatus. The study included 91 hoarse children aged 6-12 (study group) and 243 healthy children (control group) of the same age. The study group underwent detailed medical history, phoniatic examination, larynx fiber endoscopy, allergy treatment, audiologic treatment, and pulmonary treatment. Pediatric Voice-Related Quality of Life questionnaire, Serbian version (PVRQOL) was completed by parents of both groups of children. We did not find statistically significant differences in the hoarse children based on diagnosis (muscle tension disorder and vocal fold nodules) and age in physical domain, socio-emotional and global domain score ($p > 0.01$). The results showed that parents did not recognize hoarseness as a health problem in children. There were significant gender differences in the group of children with hoarseness, i.e. parents in all three PVRQOL questionnaire domains recognized hoarseness as a significant health problem in girls, but not in boys. The presence of hoarseness impairs the quality of life in pediatric population. Social and emotional domains indicated greater impact in boys.

Key words: *Children; Dysphonia; Quality of life; Pediatric Voice Related Quality of Life questionnaire*

Introduction

Instruments that measure quality of life should be able to assess physical, social and emotional issues and contain several domains in the Pediatric Voice-Related Quality of Life (PVRQOL) protocol not only providing a total score, but also the physical and socio-emotional dimensions. Voice is essential for interpersonal relationships and good verbal communication, and any changes in its production or quality may trigger organic, social, emotional, and even academic disadvantages¹. From the earliest stages of life, the closest

family members and social environment (peer groups, nurseries, kindergartens, and schools) play a major role in the formation of the child's voice.

Hoarseness occurs in children of both genders, from the earliest age and beyond, and is caused by improper use or overuse of vocal apparatus. In their daily environment, children tend to stress vocal apparatus, speaking in raised voice without any control, yelling, screaming, which results in hoarseness²⁻⁵. Children are usually treated by phoniaticians, speech therapists, due to the stress while speaking or long-term hoarseness.

Vocal fold nodules are the most common organic causes of pediatric hoarseness^{4,5}. In addition to chronic phonotrauma, which is the main etiologic factor of vocal fold nodules, specific psychological characteristics of children and their behavioral expression (dominat-

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ing their environment, talking too loudly, especially in extrovert children) are usually listed⁶.

Irregular phonation automatisms are difficult to dissolve, thus long-term vocal therapy is required for the installation and stabilization of new regular conditional reflexes of vocal hygiene.

Subjective parental evaluation of voice quality on the questionnaire scale is a reliable tool used to assess quality of life in adults in the sociological, emotional and physical function domains (Voice Related Quality of Life-In-RQOL)². Since 2001, it includes functional assessment of voice pathology by the European Laryngological Society⁴. This questionnaire was further developed for use in children with a variety of disorders including voice problems and is completed by parents assessing the quality of life in hoarse children³.

A series of surveys assessing quality of life as a parameter for the impact of disease on daily life of patients with hoarseness have emerged during the last decade. It has been indicated that voice assessment in hoarse children is a good parameter referring to the severity of hoarseness. Accordingly, pediatric and standardized questionnaires measuring the quality of life in children with hoarseness have also been designed⁷⁻⁹.

The PVRQOL questionnaire is a valid instrument evaluating the quality of life in hoarse children and relies on the parent report as a source of information. The questionnaire includes physical, socio-emotional and functional aspects of hoarseness³.

In Serbia, hoarseness in children has not been sufficiently recognized as a health problem that needs to be treated, which if not treated, leads to numerous consequences in the population of preschool and schoolchildren. Therefore, the objective of our study was to determine the quality of life in hoarse children aged 6-12 years by using PVRQOL questionnaire completed by parents.

Subjects and Methods

Cohort studies that involved pediatric hoarse patients were performed at the Phoniatic Division, Otorhinolaryngology Department (tertiary institution), Kragujevac Clinical Center in Kragujevac, during a 3-year period (2011-2014). The study included 91 hoarse children (study group) aged 6-12 and control group of 243 age-matched healthy children from

two elementary schools in two Serbian towns. All subjects gave their informed consent for participation in the study and ethical guidelines of the Declaration of Helsinki were followed during the study. Healthy children were chosen by the method of randomized sample in two elementary schools. Study group underwent detailed medical history, phoniatic examination, larynx fiber endoscopy, allergy treatment, audiologic treatment, pulmonary treatment, and multidimensional computer software voice analysis (jitter, shimmer and standard deviation). The study applied PVRQOL questionnaire as a valid instrument for assessing life quality, which was filled-out by the parents of hoarse children and parents of healthy children. The questionnaire was translated from English in Serbian language and culturally adapted for each item. To ensure accuracy of the translated questionnaire, back translation to English was done by another translator who was unfamiliar with the original version. The questionnaire was administered by the first author through face-to-face interviews. The PVRQOL is a 10-item instrument designed to measure Voice-Related Quality-of Life (VRQOL) adapted from the adult VRQOL instrument. The scores of the instrument have been normalized to a scale of 100 for ease of interpretation. A score of 100 represents the highest QOL, meaning that the parents perceived no problems with their child's voice, no limitations on voice function, and no adverse social or emotional effects attributable to their child's voice quality. In the hoarse children, diagnosis had already been made during initial examination and appropriate therapy determination. However, the study did not include treatment results, as the goal of the study was assessment of the quality of life in hoarse children at first examination.

Complete statistical analysis was performed using IBM SPSS Statistics 19.0 computer program. All continuous variables (age, scores of scales) were expressed as mean \pm standard deviation, and categorical variables (gender, marital status) as percentage of particular category frequency. For categorical variables, statistical significance of differences was assessed by χ^2 -test, while differences in continuous variables were tested by Student's *t*-test for independent causes or Mann-Whitney U test. Correlation between two continuous variables was examined by Pearson linear correlation or Spearman rank correlation.

Results

The study involved 91 hoarse children (study group) aged 6-12 and control group of 243 age-

Table 1. Percentage distribution of disorders diagnosed in study group

Disorder	n	%
Muscle tension disorder	63	69.2
Vocal cord nodules	28	30.8

Table 2. Patient demographic characteristics

	Number (%)		p value
	Hoarse subjects, 91 (26.8)	Control subjects, 248 (73.2)	
Gender*:			
male	60 (65.9)	122 (49.2)	p=0.009***
female	31 (34.1)	128 (50.8)	
Age, yrs (mean±SD)**	8.64±1.79	8.79±1.60	p=0.141
Age, yrs (mean±SD)**:			
male	8.53±1.89	8.75±1.65	p=0.179
female	8.84±1.59	8.83±1.56	

* χ^2 -test; **Mann-Whitney test; ***statistically significant

matched healthy children. None of the study group children were born from multiple pregnancies and only five children were delivered by cesarean section. Children were of regular psychomotor development according to gender and age. All children had normal audiology findings. Study group mostly included families with two children (n=50; 54.9%). There were 60 (65.9%) male and 31 (34.1%) female children, mean age 8.64±1.79 years. Table 1 shows distribution of disorders diagnosed in the study group. Muscle tension disorder was present in 63 (69.2%) and vocal fold nodules were detected in 28 (30.8%) children. Clinical examination and fiber laryngoscopy revealed that all children had pronounced outward signs of hyperkinesia and insufficient glottic occlusion with sound in the speech voice. Allergic rhinitis as comorbidity was found in 13 (14.3%) and bronchial asthma in seven (7.7%) children. There was no statistically significant age difference between the two groups (p<0.01) but there was a statistically significant gender difference (Table 2). We analyzed computerized voice parameters in relation to age and gender in the study group that included patients divided by age into two subgroups of 6-8 and 9-12 years. There were no statistically significant differences between study subgroups in jitter and shimmer according to gender and age (p>0.01). Our results of PVRQOL questionnaire suggested that par-

Table 3. Mean values of all scale scores in two groups (PVRQOL)

Question	Hoarse subjects		Control subjects		p values
	Mean	SD	Mean	SD	
Item 01	2.14	1.16	1.31	0.79	p<0.001*
Item 02	1.40	1.02	1.18	0.61	p=0.392
Item 03	1.13	0.34	1.10	0.41	p=0.111
Item 04	1.13	0.65	1.33	0.73	p=0.001*
Item 05	1.26	0.68	1.09	0.40	p=0.024
Item 06	1.00	0.00	1.08	0.33	p=0.021
Item 07	1.14	0.35	1.05	0.27	p=0.001*
Item 08	1.00	0.00	1.04	0.24	p=0.066
Item 09	1.29	0.50	1.12	0.44	p<0.001*
Item 10	1.00	0.00	1.01	0.14	p=0.391
Physical domain score	8.10	2.57	6.83	1.93	p<0.001*
Socio-emotional domain score	4.40	0.91	4.48	1.16	p<0.001*
Global domain score	12.49	3.25	11.31	2.88	p=0.429

*statistically significant (χ^2 -test); PVRQOL = Pediatric Voice-Related Quality of Life questionnaire

Table 4. Gender differences in scale scores in two groups (PVRQOL)

		Hoarse subjects		Control subjects		p values*
		Mean	SD	Mean	SD	
Physical domain score	Male	7.03	1.31	6.67	1.61	p=0.001
	Female	10.16	3.12	6.99	2.19	p<0.001
	p values**	p<0.001*		p=0.290		
Socio-emotional domain score	Male	4.07	0.36	4.48	1.17	p=0.001
	Female	5.03	1.25	4.48	1.14	p=0.005
	p values**	p<0.001*		p=0.807		
Global domain score	Male	11.10	1.41	11.15	2.63	p=0.029
	Female	15.19	4.04	11.47	3.10	p<0.001
	p values**	p<0.001*		p=0.401		

*Statistically significant; **Mann-Whitney test; PVRQOL = Pediatric Voice-Related Quality of Life questionnaire

Table 5. Voice-Related Quality of Life (V-RQOL) scores according to subject groups

Quiz score	V-RQOL score	Hoarse subjects	Control subjects	p value*
10	100 (excellent)	28 (30.8%)	166 (66.9%)	<0.001
20	75 (fair to good)	63 (69.2%)	76 (30.6%)	
30	50 (poor to fair)	-	5 (2.0%)	
40	25 (poor)	-	1 (0.4%)	
50	0 (worst possible)	-	-	

* χ^2 -test

ents did not recognize hoarseness as a health problem in children, nor that a raised tone of voice, yelling and vocal fold strain was related to hoarseness. Hoarseness in children was associated with significantly high parental response to item 01 (“My child has trouble speaking loudly or being heard in noisy situations”), item 07 (“My child has trouble doing his or her job or schoolwork because of his or her voice”) and item 09 (“My child has to repeat himself or herself to be understood”) of PVRQOL questionnaire (Table 3). In the study group (hoarse children) we found that allergic rhinitis in children affected the significantly high parental response to item 01 (“My child has trouble speaking loudly or being heard in noisy situations”), item 07 (“My child has trouble doing his or her job or schoolwork because of his or her voice”) and item 08 (“My child avoids going out socially because of his or her voice”) of PVRQOL questionnaire (χ^2 -test; $p<0.01$). On the other hand, asthma in children was associated with the significantly high parental response to item 01 (“My child has trouble speaking loudly or being heard in noisy situations”), item 05 (“My child some-

times gets depressed because of his or her voice”) and item 07 (“My child has trouble doing his or her job or schoolwork because of his or her voice”) of PVRQOL questionnaire (χ^2 -test; $p<0.01$). Using the same test, we established that gender did not affect only item 08, while the number of children in the family did not affect only item 09 (“My child has to repeat himself or herself to be understood”) of PVRQOL questionnaire (χ^2 -test; $p<0.01$). Analysis of gender differences in scale scores between the two groups showed that there was a statistically significant gender difference when looking at the group of hoarse children, i.e. parents in all three domains recognized hoarseness as a significant health disorder in girls. On average, the scores were significantly higher in girls (Table 4). We did not find statistically significant difference in hoarse children according to diagnosis (muscle tension disorder and vocal fold nodules) and age in the physical, socio-emotional and global domain scores ($p>0.01$). Based on the distribution of frequencies and absolute numbers, hoarse children had the highest frequency rate with score 20, that is fair to good, whereas in the con-

control group the highest frequency rate had score 10, i.e. excellent, which means that the questionnaire is good and that it showed lower values in the group of hoarse children than in control group. On the other hand, there were children with a score of 30 and 40 in the control group, indicating that parents failed to recognize that these children had a voice problem (Table 5).

Discussion

Pediatric dysphonia is characterized as any difficulty that hinders or prevents children from producing their natural voice. Vocal changes during childhood negatively interfere with the social, affective, and emotional development of children. The impact of vocal problem on the life of children may be underestimated, considering that they do not present more comprehensive symptoms involving other systems¹⁰⁻¹².

In order to assess the quality of life in hoarse children, hetero history, clinical and otorhinolaryngologic examinations, as well as acoustic perception are not sufficient. There is only one instrument that measures the voice-related quality of life in pediatric population, and it is a questionnaire for parents. The reference literature data confirm the relevance of this research^{3,13}.

Our research included children aged 6-12, mean age 8.64 years. Boseley *et al.* included children aged 2-18 years in their research; there were more boys than girls³. Our study results are consistent with the literature suggesting more frequent occurrence of vocal changes in the 5- to 10-year age groups, more present in boys than in girls. The prevalence of pediatric dysphonia in schools varies from 6% to 23.4%, reaching its peak in children aged 5-10^{1,3,6,9,14}.

The questionnaire used in our study was adapted in accordance with Boseley *et al.* PVRQOL validation, taking into account the fact that each society exists in the context of different cultures, beliefs, habits, attitudes and behavior patterns, which influence the perception of vocal changes, as well as the search for appropriate research and treatment³.

Given the aforementioned, the research included translation and back-translation of PVRQOL as to confirm whether the subjects had difficulties in understanding the segments of the instrument used, as well as to test the questions which are not applicable in the context of Serbian culture. The translated version was thus applied to the group of 20 subjects, enabling the

option 'unclear' for all of the questions that were not clear enough and applicable for the domain being evaluated. Only appropriately adjusted questionnaire could be applied to a wider sample as ours⁹.

Vocal assessment in children has characteristics different from those in adults, such as little cooperation in tests, small-sized laryngeal structures, and vocal similarities between genders in preschool and school-children, change of vocal parameters near the time of voice change, and difficulties in defining normative parameters for vocal samples in different age ranges. Our study group was divided into two age groups of 6-8 and 9-12 years according to the intensity of hoarseness. We noticed that the latter age group (9-12 years) involved children with more intense phonation automatism.

Results of multidimensional computer software voice analysis (jitter, shimmer and standard deviation) yielded no statistically significant difference in the homogeneous population. In the older group (9-12 years), the values were higher for each of the three parameters but without statistically significant difference ($p>0.01$). These findings were expected, as children of that age are already on the verge of puberty, when differences between the genders are becoming more prominent¹⁵.

On a large sample of 312 children, Angelillo *et al.* demonstrated that hoarseness is most common between the age of 2 and 16, with a large prevalence among boys as compared to girls¹⁶. Similar were our findings that indicated a higher prevalence in male children ($n=60$; 65.9%) than in female children ($n=31$; 34.1%). In our study, significant gender differences were also found when observing the group of hoarse voice children, i.e. hoarseness was more frequently recognized in girls as a significant health problem in all three domains by parent reports. Families with two children predominated in our study ($n=50$; 54.9%). In the study conducted by Angelillo *et al.*, families with more than two children were dominant, which can be explained by the size of the test sample. It has been noticed that vocal abuse in children occurs when extrovert children want to dominate their environment. In this study, there were 79% of extrovert children, while the study conducted by Italian authors included 83% of such children¹⁶.

Walz *et al.* carried out a study to determine the risk factors for hoarseness in premature infants. Literature

data suggest that prolonged intubation is associated with poorer long-term parent-perceived voice quality in these patients¹⁷. In our study group, most children were born without complications. Only five deliveries were completed by cesarean section. There were no premature infants.

In a large retrospective study of pediatric hoarse voice patients, Shah *et al.*¹⁸ found that muscle tension disorder correlated with vocal fold nodules, which was confirmed by our research. Muscle tension disorder was found in 63 (69.2%) and vocal fold nodules in 28 (30.8%) children. We also detected comorbidities of allergic rhinitis in 14.3% and asthma in 7.7% of the study group of children. Allergic rhinitis affected the function of voice resonator, referring to the parental statistically significant responses to items 01, 07 and 08 in PVRQOL questionnaire. All children with allergic rhinitis were of the extrovert type. Jitter values and shimmer vocal assessments in this group were slightly higher than the normal range. In our study group, seven (7.7%) patients had bronchial asthma, which influenced statistically significantly responses to items 01, 05 and 07 in PVRQOL questionnaire. This finding is expected especially in children with a longer period of inhalation therapy (pump)¹⁹.

The extrovert quality in these children was established by using hetero history records provided by the parents or custodians. Bronchial asthma and use of inhaled medications additionally disrupt vocal function voice activator, which was confirmed by vocal assessment. PVRQOL as an instrument directly leaning on parent responses is used to determine how hoarseness affects the quality of life in their children^{3,10,19}.

Connor *et al.* conducted a study on dysphonic children aged 5-18 years, which showed a discrepancy between the children's and their parents' reports on their voices, with a conclusion that children at an early age were able to comment their vocal disorders. Their study also suggested that chronic hoarseness in children negatively affected the quality of their life¹².

Verduyck *et al.* have indicated that subjective evaluation of voice is routinely used in adults, but is not appropriate for children. Therefore, they used a questionnaire for children and their mothers in voice self-assessment. They concluded that children could express voice disorders, although they defined them differently than their parents or custodians¹⁵.

Tavares *et al.* examined hoarse voice children with nodules and made comparisons with the control group

of children without vocal disorders by employing vocal self-assessment methods and multi-dimensional computerized voice analysis. It was observed that some parents often overestimated hoarseness in children, while others did not consider hoarseness as an important symptom, giving biased answers to the questions assessing the quality of life, thus potentially causing delay in reaching an accurate diagnosis^{20,21}.

Timely identification of improper voice use and voice misuse can lead to significant reduction in the prevalence of voice disorders in children, i.e. improvement in the children's quality of life.

So far research has shown that younger children are not mature enough, nor do they possess cognitive abilities for self-assessment, and therefore questionnaires for parents or custodians are necessary to assess the child's vocal status^{4,7-10}.

However, older children are capable of self-assessment using adapted questionnaires. By applying different assessment instruments, in our study the PVRQOL scale, clinicians and researchers can monitor progression of the disorder, as well as efficiency of the treatment.

In conclusion, taking all this in consideration, it is necessary to educate parents and teachers in order to attain proper vocal hygiene and appropriate voice production. Likewise, it is of great importance that healthcare professionals such as general practitioners and pediatricians, who come first in contact with the hoarse voice child, be acquainted with the issue. The PVRQOL questionnaire demonstrated that hoarseness did not affect the quality of life in the children, as indicated by the global domain score of the study children.

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Sažetak

KVALITETA ŽIVOTA PROMUKLE DJECE U SRBIJI
MJERENA UPITNIKOM *PEDIATRIC VOICE-RELATED QUALITY OF LIFE* (PVRQOL)

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Promuklost se javlja kod djece oba spola, od najmlađe dobi i kasnije, a uzrokovana je nepravilnom upotrebom ili zloupotrebom vokalnog aparata. Ova studija obuhvatila je 91 promuklo dijete u dobi od 6-12 godina (ispitivana skupina) i 243 zdrave djece iste dobi (kontrolna skupina). U ispitivanoj skupini učinjena je detaljna anamneza, fonijatrijski pregled, fiberoskopija larinksa, alergološki tretman, audiološki tretman i pulmološki tretman. Roditelji djece obiju skupina odgovorili su na upitnik *Pediatric Voice-Related Quality of Life* (PVRQOL) na srpskom jeziku. Nije pronađena statistički značajna razlika kod promukle djece na osnovi dijagnoze (hiperkinetska disfonija i noduli glasnica) i dobi u fizičkoj domeni, u socio-emocionalnoj domeni i u globalnoj domeni ($p > 0,01$). Rezultati su pokazali da roditelji nisu prepoznali promuklost kao zdravstveni problem kod djece. Postoje značajne spolne razlike u skupini promukle djece, tj. roditelji su prepoznali promuklost kao značajan zdravstveni problem u sve tri domene upitnika PVRQOL kod djevojčica, ali ne i kod dječaka. Prisutnost promuklosti smanjuje kvalitetu života u pedijatrijskoj populaciji. Socijalna i emocionalna domena imale su veći utjecaj kod dječaka.

Ključne riječi: *Djeca; Promuklost; Kvaliteta života; Upitnik Pediatric Voice Related Quality of Life*